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Case Report

Deep-infiltrating endometriosis with primary presentation as obstructive uropathy: report of two cases

Richa Vatsa¹, Neena Malhotra¹, Smita Manchanda², Juhi Bharti^{1*}

¹Department of Obstetrics and Gynaecology, All India Institute of Medical Sciences, New Delhi, India

²Department of Radiology, All India Institute of Medical Sciences, New Delhi, India

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*Correspondence:

Dr. Juhi Bharti,

E-mail: drjuhigeorgian.04@gmail.com

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ABSTRACT

Endometriosis can have a broad spectrum of presentation and may sometimes affect/encase ureters leading to scarring, adhesion formation and obstruction resulting in backpressure changes in kidneys and ureters. Ureteral endometriosis is generally asymptomatic for long term. By the time symptoms arise, renal damage can be irreversible. Two of our cases had an unusual presentation of flank pain, mimicking urological pathology. Both had obstructive uropathy without any of the typical gynecological features of endometriosis like dysmenorrhea, chronic pelvic pain or infertility. One had a unilateral nonfunctional kidney due to long standing hydroureteronephrosis, for which a nephrectomy was done. In other, obstructive uropathy was managed with bilateral percutaneous nephrostomy. Fertility-preserving surgery for endometriosis was done in both the cases. To conclude, endometriosis can have non-gynecological presentation also. So, in reproductive age females presenting with obstructive uropathy, endometriosis should always be kept as differential diagnosis. Endometriosis can be a silent killer for kidneys.

Keywords: Deep infiltrating endometriosis, Nephrectomy, Obstructive uropathy

INTRODUCTION

Urinary tract involvement by endometriosis has been seen in 0.3-12% of all cases of endometriosis out of which bladder endometriosis is the most common (85%) followed by that of ureter (9%), kidneys (4%) and urethra (2%).¹⁻³

Endometriotic lesions encasing ureters may lead to scarring and adhesion formation. These adhesions can cause ureteral obstruction resulting in backpressure changes in kidneys and ureters.⁴

We are reporting two unusual cases of renal damage caused by deep infiltrating endometriosis and surprisingly there were no gynecological symptoms of endometriosis in both of these cases.

CASE REPORTS

Case 1

A 27-year-old female presented with a complaint of severe pain in left loin for 10 months. On probing, she revealed a history of spasmodic dysmenorrhea for last 15 years which was not bothersome and hence, she did not consult any doctor. She had been married for 10 months, without any history of infertility.

She had a history of Double J (DJ) stent insertion for obstructive uropathy, which was removed 3 months later. She was prescribed hormonal medications for endometriosis diagnosed during a workup for pain in the abdomen. Ultrasound (USG) showed grade IV and I hydroureteronephrosis (HDN) of the left kidney and right kidney, respectively. The left adnexa had a 62×33 mm

endometriotic cyst in left ovary along with hydrosalpinx. similarly, there was a 75×51 mm endometriotic cyst along with hydrosalpinx in right adnexa. Uterus had posterior wall adenomyoma of 2.9×3 cm. Her blood investigations showed normal kidney function test (KFT). Her CA-125, CEA and CA 19.9 values were 196 U/ml, 1.13 ng/ml and 63.94 U/ml respectively. She had decreased ovarian reserve also, her AMH was 0.22 ng/ml. Contrast-enhanced Computed tomography (CECT) scan revealed bilateral endometriotic cysts with left gross hydroureteronephrosis and mild right-sided hydroureteronephrosis (Figure 1 A-F).

The DTPA scan showed non-visualization of left kidney along with normal functioning right kidney. Surgery was done by joint team of gynecologists and urologists. Intraoperatively there were bilateral endometriotic cysts of 4x3 cm size along with bilateral hydrosalpinx. Bilateral ovaries were adherent to ovarian fossa and there were dense adhesions between sigmoid colon and posterior surface of uterus. Left kidney and ureter, both were grossly dilated (Figure 2A).

Laparoscopic left nephrectomy and cystoscopy guided right-sided DJ stenting, bilateral ovarian cystectomy and bilateral salpingectomy were done. Later levonorgestrel-releasing intrauterine system (LNG-IUS) was inserted for prevention of recurrence. Histopathology confirmed endometriosis. Patient has been under follow-up for last 1 year and doing well.

Case 2

A 27-year-old unmarried female had complained of pain in her left loin and abdominal swelling on her left side for the last 2 months. The pain was relieved with oral analgesics. She presented to our emergency with sudden onset severe pain in her left loin. She did not have any menstrual complaints. Emergency USG reported grade IV HDN in bilateral kidneys, ovarian endometrioma of size 12.5×5.5 cm on the left side and 12×6 cm on the right side. She had deranged kidney function with serum creatinine as 2.6 mg/dl; therefore, it was decided to proceed for emergency bilateral percutaneous nephrostomy (PCN). Her creatinine value improved to 1.1 mg/dl, one month after PCN insertion. Her CA-125, CEA and CA 19.9 values were 73.3 U/ml, 2.02 ng/ml and 1.2 U/ml, respectively.

With the provisional diagnosis of endometrioma causing pressure effect on bilateral ureters, she underwent surgery. Intraoperatively, there was a 10x8 cm ovarian endometrioma with pus on the left side and a 10 x 12 cm endometrioma on the right side (Figure 2B). The pouch of Douglas was completely obliterated. She underwent laparoscopic left salpingo-ovariotomy and right ovarian cystectomy.

Bilateral PCN was removed 2 weeks after surgery. The patient has been asymptomatic after surgery without any evidence of hydroureteronephrosis.



Figure 1: CECT images of bilateral endometriomas with ureteric involvement. (A and B) Axial images reveal thick-walled, multilocular cystic lesions (* in A) in the bilateral adnexa. Bilateral ureters are coursing posteriorly (arrows in B); (C and D) coronal reformatted images reveal left gross hydroureteronephrosis with marked thinning of the renal parenchyma (arrows). The right ureter is also dilated (dotted arrow); (E) (coronal MIP) and (F) VRT images of CECT in the delayed phase reveal mild right hydronephrosis and hydroureter (arrow) with narrowing of the ureter at the level of the right endometrioma (dotted arrow). The left kidney is poorly excreting with no contrast seen in the left pelvicalyceal system in the delayed phase (*).

Table 1: Investigation of both patients.

	Case 1	Case 2
Serum urea	23	17
Serum creatinine	0.7	2.6 (before DJ* stent), 1.1 (a month after DJ* stent)
Seum AMH	0.22	Not done
CA 125	196	73.3

Continued.

	Case 1	Case 2
CEA	1.13	2.02
CA 19.9	63.94	<2
USG	Left kidney: grade IV HDN†, Right kidney: grade 1 HDN†, Left adnexa: 62×33 mm endometriotic cyst along with hydrosalpinx, Right adnexa: 75×51 mm endometriotic cyst along with hydrosalpinx, Uterus: posterior wall adenomyoma 2.9×3 cm	B/L kidneys grade IV HDN†, Ovarian endometrioma of 12.5×5.5 cm on left side and 12× 6 cm on right side.

* Double J, † Hydronephrosis

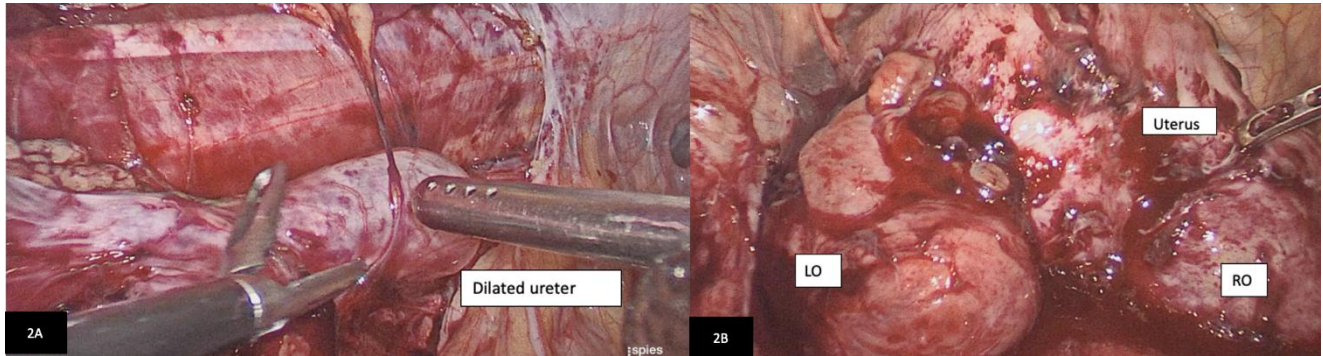


Figure 2: (A) Intraoperative picture showing grossly dilated ureter; (B) Intraoperative picture showing bilateral tubo-ovarian masses.

DISCUSSION

Ureteral endometriosis is generally asymptomatic in terms of urological symptoms for long term as seen in both of our cases. By the time symptoms arise, renal damage may become irreversible necessitating nephrectomy; similar to first case reported here. Both of our cases presented with severe obstructive uropathy.

It can be a silent killer for kidneys, as per an estimate around 25-50 % nephrons are lost and 30% cases of ureteral endometriosis will have loss of renal function by the time of presentation.⁵ Though on history taking long standing primary dysmenorrhea was revealed on probing in one case, but that was not presenting feature.

On workup of obstructive uropathy, primary pathology was endometriosis in both cases causing mass effect on ureters. Ureteric involvement in endometriosis can be either due to external compression or transmural implantation. In both of cases, external compression was main pathology and after adhesiolysis, it was possible to save the ureters.

Similar to our cases, Alaoui et al reported case of 42-year-old female who presented with flank pain without any gynaecological complain.⁶ Imaging detected unilateral hydronephrosis due to endometrioma and ureteric endometriosis. Patient underwent laparotomy, resection of diseased ureter and anastomosis.

Similarly, Tanaka et al and Mason et al reported cases with non-gynaecological primary presentation in cases of

endometriosis, workup of which revealed unilateral HDN with loss of renal function, both managed with unilateral nephrectomy.^{7,8} Majority of cases of UTE reported in literature, patients generally have at least one symptom of endometriosis like CPP, dysmenorrhoea, infertility etc. Ota et al, reported a case of 49-year-old woman presenting with chronic pelvic pain.⁴ On workup, endometrioma with deep infiltrating endometriosis (DIE) was detected, causing bilateral ureteral obstruction and loss of renal function. This was managed with ureterolysis. Whereas

Choi et al reported a case of acute renal failure due to obstructive uropathy leading to unilateral nephrectomy. The patient was 45-year-old post hysterectomy, which was done 6 years ago along with salpingectomy for severe dysmenorrhoea. She had recurrence of endometriosis in left hemipelvis along with HDN and loss of renal function on that side.⁹ Talreja et al, presented three cases of ureteral endometriosis, presentations related to infertility, dysmenorrhoea, dyspareunia and chronic pelvic pain in various combinations.¹⁰ All were managed with laparoscopic ureterolysis and endometriotic cystectomy with or without DJ stenting.

Ureteric involvement in endometriosis can be either due to external compression or transmural implantation. In both of our cases, external compression was the main pathology. Management of obstructive uropathy may vary from ureterolysis to nephrectomy, depending on the degree of involvement at the time of presentation. A non-functioning kidney needs to be removed as it can result in recurrent pyelonephritis or stone formation.

Wang et al, did a retrospective study on 82 patients of urinary tract endometriosis (UTE) and concluded that diagnosis of ureteral endometriosis requires high index of suspicion.¹¹ Medical management can be tried in patients of UTE in the absence of hydronephrosis. Surgical management is the treatment of choice in cases of no response to medical management or hydronephrosis as delayed management may lead to irreversible loss of renal function.¹² The patients undergoing non-surgical management of endometriosis, regular USG can be done to rule out development of obstructive uropathy.

CONCLUSION

Obstructive uropathy due to endometriosis can damage kidneys, and if not treated on time, can even result in complete loss of renal function. Urinary tract symptoms due to endometriosis can be a presenting feature of endometriosis. Therefore, there should be a high index of suspicion of urinary tract endometriosis in cases presenting with obstructive uropathy in the reproductive age group without any obvious renal pathology.

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